

Selective and Superselective Angiography of Pediatric Moyamoya Disease Angioarchitecture in the Posterior Circulation

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Summary

The anastomotic network of the posterior circulation in children with moyamoya disease has not been analyzed. We aimed to investigate the angiographic anatomy of this unique vascular network in patients with childhood moyamoya disease.

Selective and superselective injections of the posterior circulation were performed in six children with newly diagnosed moyamoya disease. The arterial branches feeding the moyamoya anastomotic network, their connections and the recipient vessels were demonstrated.

Depending on the level of the steno-occlusive lesion, the feeding vessels were the thalamoperforators, the posterior choroidals, the splenic artery, parietoccipital artery, other cortical posterior cerebral artery (PCA) branches, the dural branch of the PCA, the premamillary artery and other posterior communicating artery perforators. Through connections, which are described, the recipient vessels were the striate and medullary arteries, other thalamic arteries with or without medullary extensions, the pericallosal artery, medial parietoccipital cortical branches of the PCA and the anterior choroidal artery.

High quality selective and superselective angiography helped in demonstrating the angiographic anatomy of the moyamoya posterior anastomotic network previously either vaguely

or incompletely described, as well as connections within the posterior circulation but also its relevance as a collateral to the anterior circulation.

Introduction

The steno-occlusive changes occurring in the posterior circulation in moyamoya disease are often overlooked. These changes are more commonly seen in children than in adults^{1,2}. The anastomotic network of the posterior circulation in children has so far not been analyzed. Hence we aimed to investigate the anatomy of this posterior vascular network in children with moyamoya disease.

The angiographic characteristics of moyamoya disease described so far³⁻⁹ were exclusively based on the images obtained during a selective internal carotid (ICA) or vertebral artery (VA) contrast injection. The information that can be extracted from such an angiographic investigation is limited, because the connections and territory of the arterial branches participating in the complex microanatomy of the collateral network at the base of the brain are obscured.

In this study, high-quality selective and magnified angiographic images were supplemented by several superselective microcatheter injections so that the precise angioarchitecture of the collateral circulation was clearly demonstrated. Particular focus on the less fully de-

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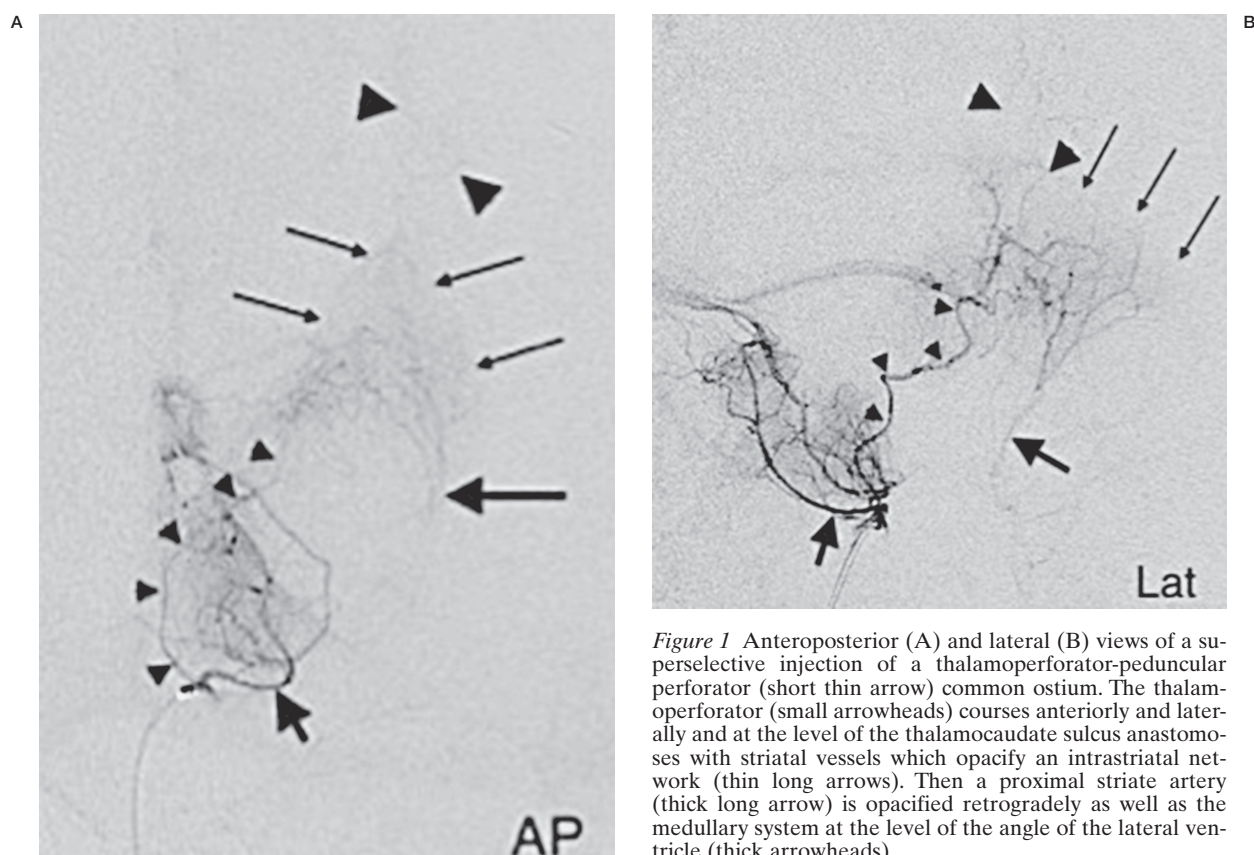


Figure 1 Anteroposterior (A) and lateral (B) views of a superselective injection of a thalamoperforator-peduncular perforator (short thin arrow) common ostium. The thalamoperforator (small arrowheads) courses anteriorly and laterally and at the level of the thalamocaudate sulcus anastomoses with striatal vessels which opacify an intrastriatal network (thin long arrows). Then a proximal striate artery (thick long arrow) is opacified retrogradely as well as the medullary system at the level of the angle of the lateral ventricle (thick arrowheads).

scribed posterior circulation network was undertaken, since it is well-known that the posterior circulation is typically less seriously affected.

Materials and Methods

During 2012, 19 children (age range from two to 13 years) diagnosed with moyamoya disease or syndrome, were hospitalized in the Moyamoya Centre, Children's University Hospital in Zurich. All children underwent digital subtraction angiography at our department. Five patients had an angiogram only after a revascularization operation with the first angiograms performed in referring institutes, whereas in 14 cases the angiogram was the initial diagnostic examination. Eight of these patients were considered moyamoya syndrome and were not included in this study. The DSA imaging of six Caucasian patients from two to 12 years of age (male:female = 3:3) with newly diagnosed moyamoya disease, were retrospectively analysed.

The present report only describes the angiographic features of the posterior circulation. The findings of the anterior moyamoya anastomotic networks have been described in a separate report.

The posterior communicating artery (Pcom) supplied either through the ICA or the basilar artery (BA) corresponds to the caudal division of the ICA and was considered part of the posterior circulation.

All six patients underwent six-vessel selective angiography. In three patients, superselective microcatheterizations (Elite 1.5F Stryker Neurovascular, Fremont, CA, USA) were performed in the posterior circulation by the first author (GB). Informed consent was obtained in all cases. The motivation-indication for superselective injections was diagnostic uncertainty about stenosis or occlusion of the posterior cerebral artery (PCA), as well as the extent of collateralization from moyamoya collaterals. Depending on the stage of angiopathy, distal arterial filling and extent of the deep and dural collaterals, the optimal planning of the number, location and type of the revascularisation pro-

cedure in all the affected arterial territories could be undertaken. The microcatheterizations were performed with the same angiographic setup, through an angiographic catheter (5Fr “Val” catheter, Cook Medical Inc., Bloomington, IN, USA) and were exclusively flow-guided with the microguidewire used only for proximal support. In none of the superselective injections was the microcatheter in a wedged position. No complications occurred.

Particular attention was paid to the identification of the individual arterial branches making up the so-called moyamoya anastomotic network, as well as their connections and direction of the blood flow. When part of the entire course and/or potential anastomoses of a dilated

arterial branch were not clearly seen, it was recorded as “not identified”.

For this retrospective study no Institutional Review Board approval was necessary.

Results

According to the Susuki classification⁴, one hemisphere was at stage 5, one at stage 4, six hemispheres were at stage 3 and four hemispheres at stage 2. Five of 12 hemispheres (in three of the six patients) showed steno-occlusive disease in the posterior circulation. Table 1 shows the identified arterial branches participating in the moyamoya anastomotic net-

Table 1 The vessels constituting the moyamoya anastomotic networks.

Vessel of origin	Identified vessel	Course	Recipient vessel	Hemispheres
P1	Thalamoperforators	Anterior and lateral	Striate and medullary arteries	F
P1	Thalamoperforators	Laterally with extensive connections to	Medullary artery	A
P1	Thalamoperforators	Posterior and lateral	Other thalamic with or without medullary artery	C,C
P2	Posterior choroidals	Choroid plexus, foramen Monroe to septum, transcallosal	Pericallosal artery	A,C,E
P2	Posterior choroidals	Choroid plexus, foramen Monroe subependymal	Striate artery	E
Distal PCA	Splenic a. and/or parietoccipital a.	Retrosplenic-posterior callosal - watershed	Pericallosal artery	A,B,B,D,D, E,E,F
Distal PCA	Other cortical PCA branches	Along the surface of the temporal and parietoccipital lobe	Mostly inferior trunk branches of MCA	A,B,B,C,C, D,D,E,E,F
Distal PCA	Dural branch of PCA	Dural vessels along the medial tentorial edge	Medial parietoccipital cortical branches PCA bilaterally	A
Pcom	Pcom perforators	Known collaterals at optic tract level 10	Ant. choroidal artery	C,C
Pcom	Premamillary a.	Wall of 3 rd ventricle, terminal sulcus, to the angle of the lateral ventricle.	Lateral striate artery at that level	B,B
Pcom	Pcom anterior perforator	Branches with anterosuperior medial course on 3 rd ventricle wall. Branches with posterosuperior lateral course to the angle of lateral ventricle.	ACA at the Acom level. Lateral striate a. and medullary arteries of MCA territory.	A
Pcom	Pcom perforators	Not identified	Not identified	C,C,D,E,E

Display of a patient's hemisphere twice (e.g. B,B) signifies the appearance of the described vessels in both hemispheres.

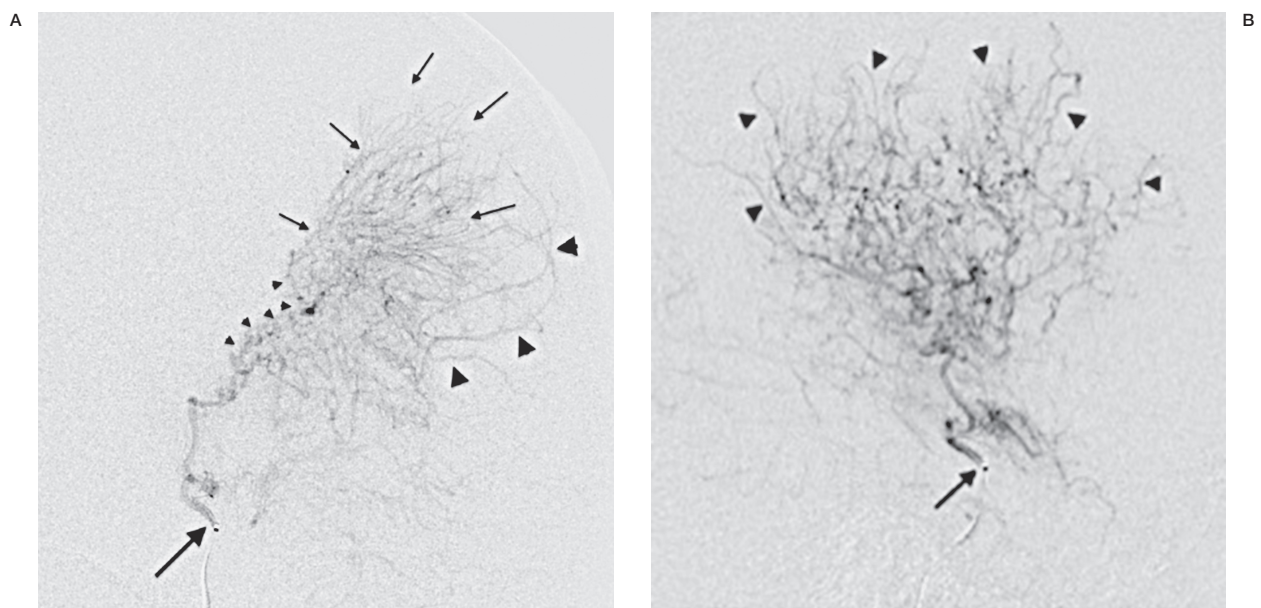


Figure 2 AP (A) and lateral (B) views of a superselective thalamoperforator injection (arrow) showing an extensive collateralization to distal MCA cortical branches (large arrowheads) with retrograde flow through medullary arteries (thin arrows) connected with the dilated thalamoperforator at the level of the lateral ventricular wall (small arrowheads).

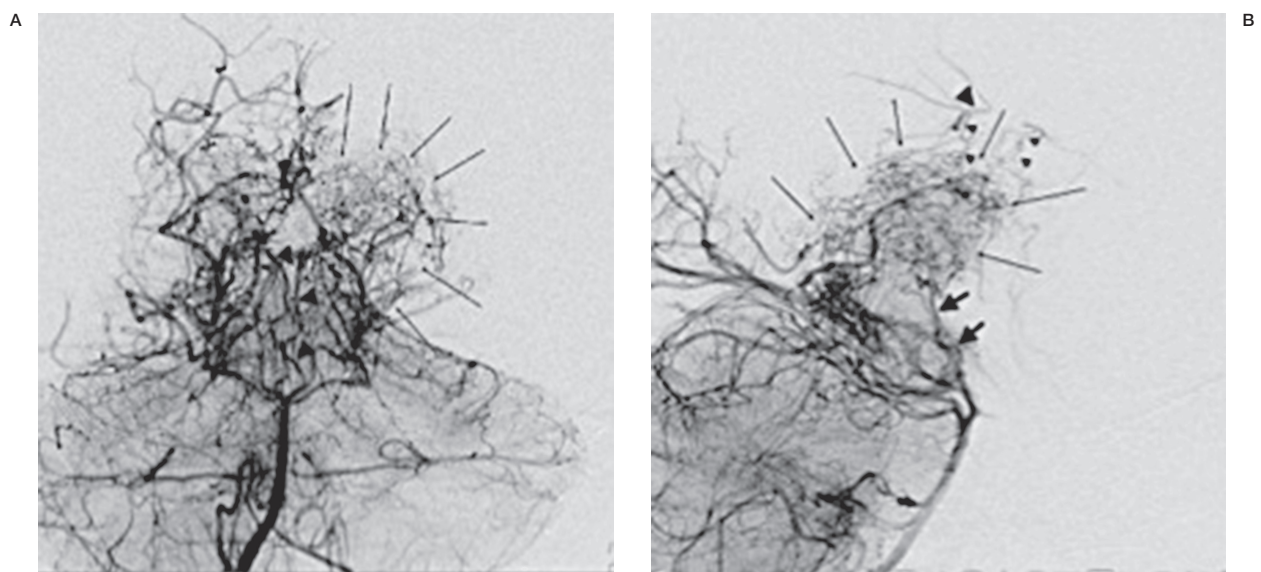


Figure 3 A) Anteroposterior view of a vertebral injection showing an intrathalamic network (long thin arrows) of dilated vessels supplied among other branches, by a left-sided thalamoperforator (arrowheads) in a patient with occlusion of the ipsilateral PCA beyond the P2 segment. B) The same injection in lateral projection showing the thalamoperforator (short arrows) connected with the intrathalamic network. The pericallosal artery (large arrowhead) is reconstructed through several septal transcortical collaterals (small arrowheads) projecting above the level of the thalamic network and most likely supplied by the midline choroidal arteries.

works, with their connections as well as the patients in which these vessels were detected.

In one case, part of the posterior moyamoya anastomotic network was constituted by thalamoperforators of the P1 segment. By su-

perselective injection, this thalamoperforator supplied lateral striate arteries and by extension medullary arteries through a relatively direct anterolateral connection (Figure 1). In another case the thalamoperforator had an ex-

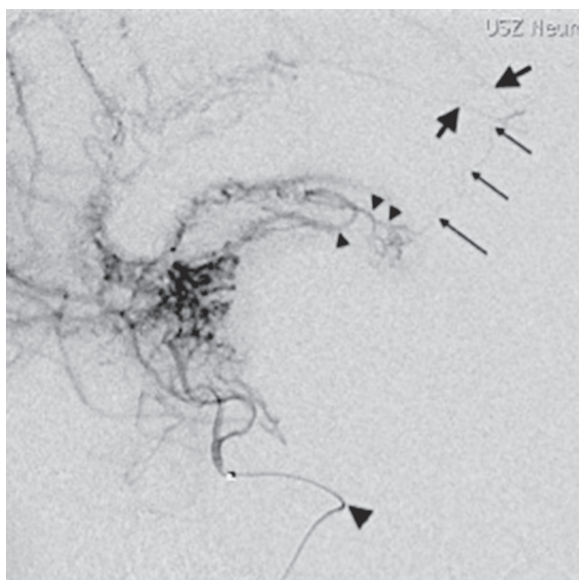


Figure 4 Lateral view of a superselective injection of the left P2 segment of another patient (large arrowhead at the basilar tip). The choroidal arteries (small arrowheads) at the level of the foramen of Monroe clearly supply septal and transcallosal arterial branches (long arrows) which reconstruct the pericallosal artery (short arrows).

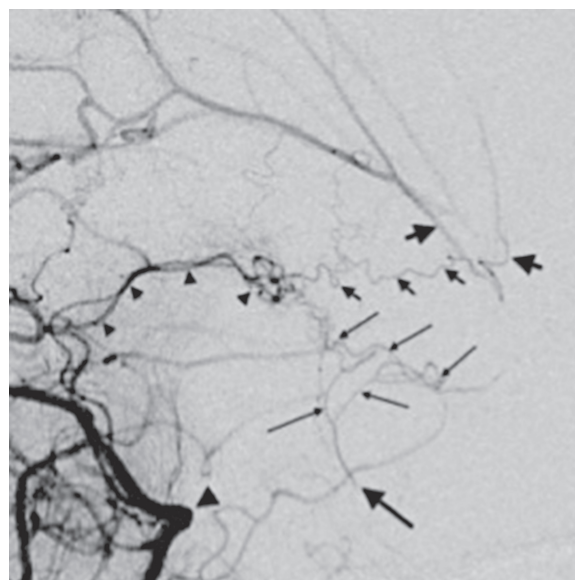


Figure 5 Lateral view of a vertebral injection showing the posterior choroidals (arrowheads) supplying both midline transcallosal branches (short arrows) which reconstruct the pericallosal artery (short thick arrows), as well as lateral branches along the floor of the lateral ventricle (thin long arrows) supplying distal striatal branches and retrogradely a single lateral striatal artery (long thick arrow).

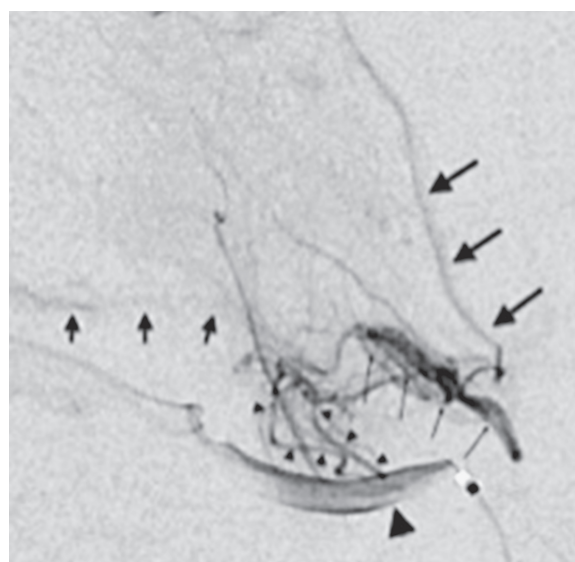
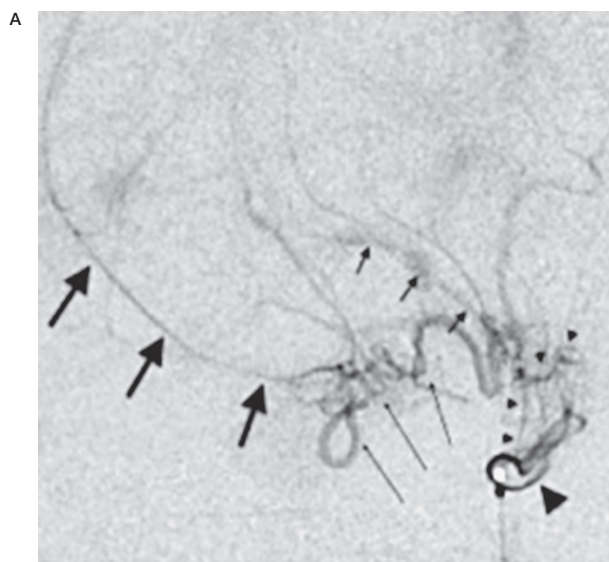


Figure 6 Anteroposterior (A) and lateral (B) views of a superselective injection of the left Pcom (thick arrowhead) which through Pcom perforators (small arrowheads) opacifies the anterior choroidal artery (small short arrows) and reconstructs the MCA (long thick arrows) via the anastomotic connection of the uncus artery (long thin arrows).

tensive contribution to the supply of the parietal MCA territory through retrograde supply of medullary arteries (Figure 2).

In a case with occlusion of the P2 segment of the PCA, the thalamoperforators of the P1

supplied the rest of the thalamus through a principally intrathalamic moyamoya anastomotic network (Figure 3).

The posterior choroidals were seen in three cases supplying the pericallosal artery through

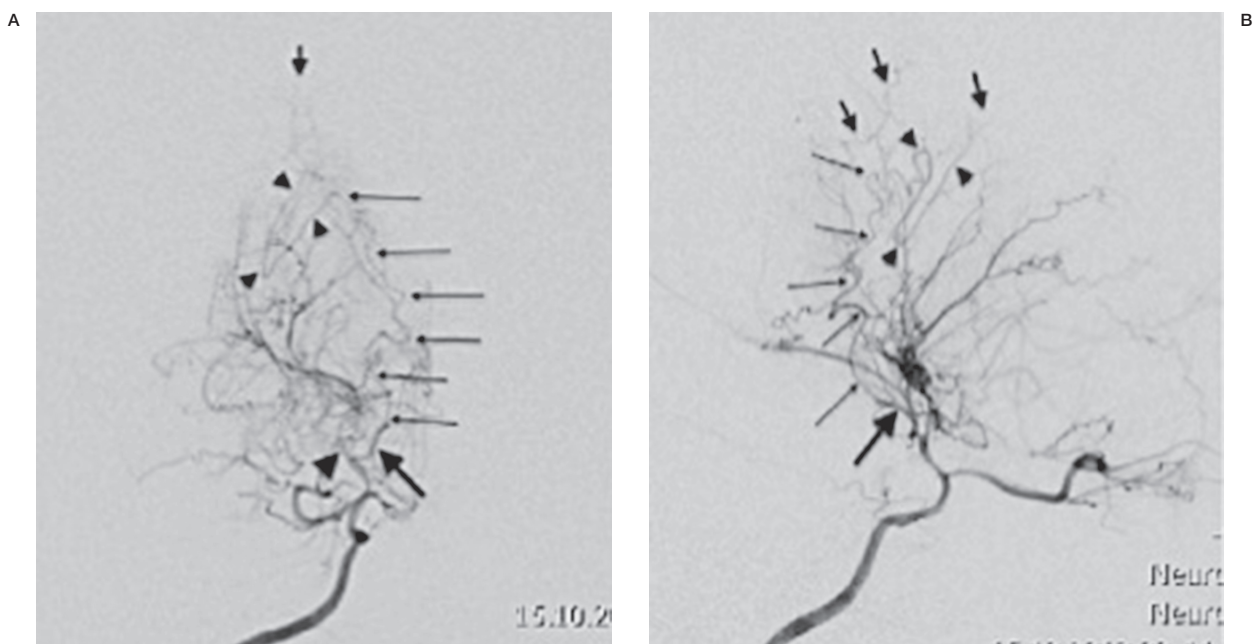


Figure 7 Anteroposterior (A) and lateral (B) views of a right ICA injection. The distal ICA (large arrowhead) is highly stenotic. The anterior choroidal is still patent with antegrade flow and the Pcom (long thick arrow) mainly supplies a dilated tuberothalamic artery (long thin arrows) which runs superiorly along the lateral wall of the posterior hypothalamus and at the level of the terminal sulcus turns lateral on the ventricular surface of the caudate nucleus where it anastomoses with the distal segment of a lateral striate artery (small arrowheads). Further opacification of the medullary system (short arrows) is better visible in the later phases (C,D).

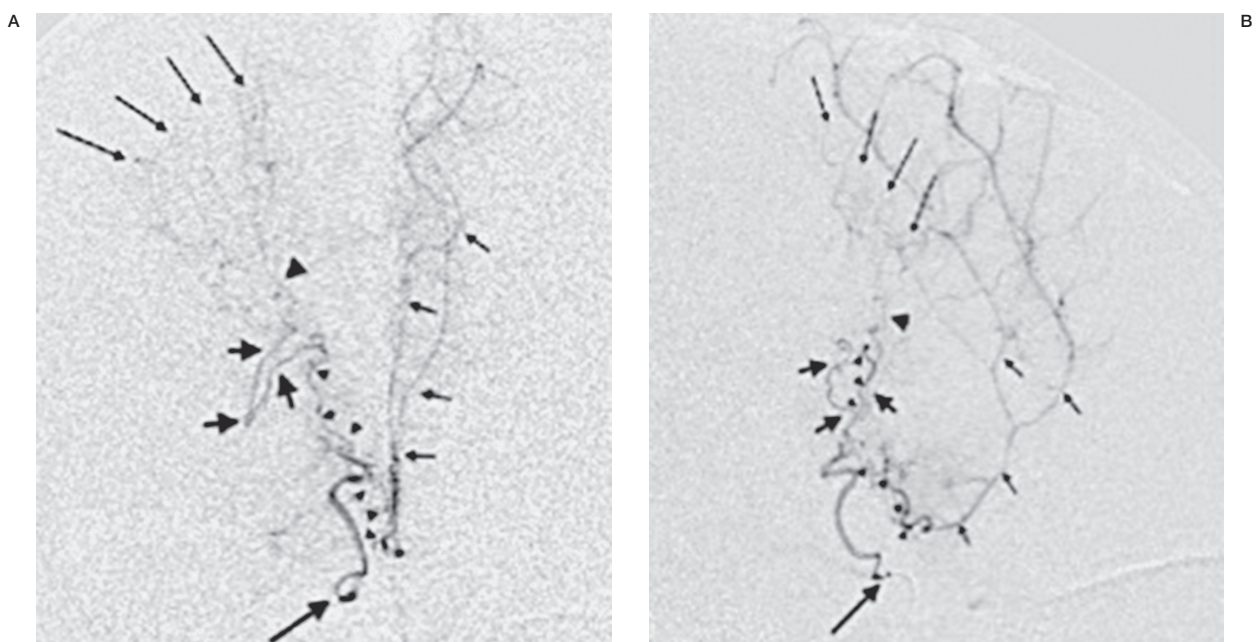


Figure 8 Anteroposterior (A) and lateral (B) views of a superselective injection of a proximal Pcom hypothalamic perforator (long thick arrow) showing collateral supply to both ACA (short thin arrows) and MCA territories. The ACA is supplied at the level of Acom with retrograde flow through hypothalamic perforators connected with the injected perforator at the level of the 3rd ventricle wall (antero-inferior arrowheads). Instead, the MCA territory is supplied with retrograde flow through the medullary arteries (long thin arrows), at the level of the angle of the lateral ventricle (thick arrowhead), fed by the distal branches of a striate artery (short thick arrows), connected (posterosuperior arrowheads) with the injected perforator.

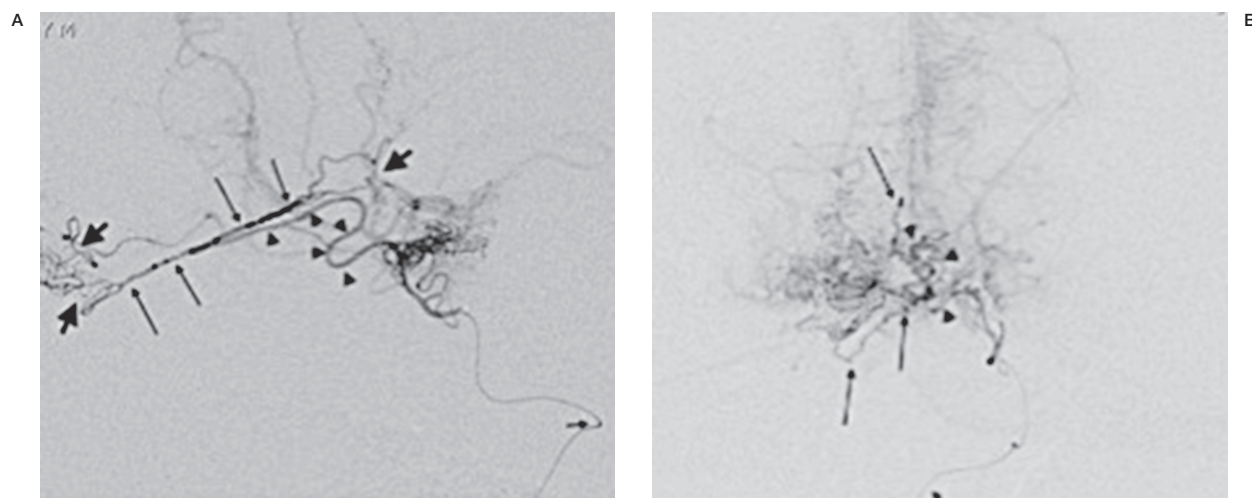


Figure 9 Lateral (A) and anteroposterior (B) views of a superselective PCA injection and its dural branch (arrowheads) which supplies another dural branch (long thin arrows) roughly parallel to the straight sinus and then through fine leptomeningeal vessels (short arrows) reconstructs the superficial medial occipital cortical arteries bilaterally. Small short arrow at the basilar tip.

midline septal and transcallosal arterial anastomotic branches at the level of the interventricular foramen (Figure 4).

In one of those cases, the choroidals through anterolateral anastomotic branches were also supplying distal striate branches reconstructing a common arterial trunk further proximally (Figure 5).

In one case of complete occlusion of the ICA at the level of the anterior choroidal, the uncus artery and its connections to the temporal branches of the MCA (described in a separate analysis of the anterior circulation) were still active due to reconstruction of the anterior choroidal artery through known Pcom collaterals (Figure 6).

In another case, the tuberothalamic artery anastomosed with the distal segment of a lateral striate artery which then irrigated retrogradely the medullary system at that level (Figure 7).

The case illustrated in Figure 8 showed a Pcom perforator being connected with both the ACA and the territory of the MCA through retrograde supply of the medullary arteries. The superselective injection of the distal PCA in a patient with occlusion of the contralateral PCA demonstrated duro-cortical anastomoses at the level of the tentorial apex which contributed to the supply of the ischemic cortex of the opposite hemisphere (Figure 9).

Discussion

While several aspects related to the angiographic features of moyamoya disease have been described^{3,4,6,8-11} the angioarchitecture of the so-called moyamoya vessels, including their microangiographic connections and direction of the flow using high quality selective and superselective angiography has never been described before. The literature to date only emphasizes their chaotic¹² nature which is frequently challenging for a detailed and meticulous analysis.

The angiographic findings of the posterior circulation in particular have attracted much less attention in the moyamoya literature than the findings of the anterior territory, presumably related to the dominant anterior circulation manifestations of the disease. In addition, since part of the posterior circulation can be visualized through an ICA injection when a prominent Pcom is present, the posterior collateral network has been underexamined.

Nashimoto et al.⁷ in 1968 included no vertebral injections in their angiographic studies. Suzuki et al.⁴ in 1969 studied 11 paediatric cases of moyamoya disease. Three of those patients had a vertebral artery injection and a follow-up angiogram. The contribution of the PCA to the moyamoya anastomotic networks was simply mentioned without detail. Handa et al.⁹ in 1972

in their angiographic study of 16 children with unspecified moyamoya did not report on the vertebrobasilar involvement due to the limited number of vertebral injections and the poor visualization of the basilar artery. Crouzet et al.⁸ in 1974 defined three types of moyamoya collaterals, all of them observed in the anterior circulation. The authors noted that the involvement of the posterior part of the circle of Willis can occur, but only rarely and later in time. Additionally they aptly noticed that the posterior cerebral arteries can supply the deep grey nuclei through their contribution to the posterior part of the moyamoya anastomotic network and through anastomoses of thalamic arteries with lateral striate arteries. Takahashi in 1980⁶, in his angiographic study of seven patients with moyamoya disease (three of them children), attempted a more detailed description of the moyamoya anastomotic networks and their interconnections. Although at least one vertebral injection was done in all cases, no particular comments on the posterior circulation were made. Miyamoto et al.¹³ in 1984 were the first to report specifically on the posterior circulation of 82 paediatric cases with moyamoya disease. They observed that the anastomotic networks are composed mainly of posterior choroidal arteries, thalamogeniculate arteries, and other thalamoperforating arteries that irrigate the thalamus and the posterior portion of the basal ganglia. When this network is well-developed it also anastomoses with medullary vessels in the parietal subcortex, but its role in providing a collateral supply was judged to be somewhat limited. Satoh et al.³ in 1988 described the angiographic findings of 34 newly diagnosed paediatric cases with moyamoya disease and distinguished the feeding branches of the moyamoya anastomotic network derived from the ICA group and PCA group. The feeding branches of the PCA group were the “premammillary, interpeduncular, thalamoperforating, thalamogeniculate perforating, medial and lateral posterior choroidal arteries”. Although it was a report with special attention to the posterior circulation angiographic changes, no further analysis of the collateral network and recipient vessels was done, and the term “abnormal net-like vessels” was used to cover this gap. In 1995, Yamada et al.¹⁴ reported on the angiographic findings of 76 patients, most of them children, emphasizing the role and changes in the posterior circulation. They referred to the leptomeningeal collaterals of the PCA to the ipsilateral ACA and MCA

territories and the posterior meningeal artery was also mentioned as a transdural contributor coming from the vertebral system. Moreover they noticed that the basal cerebral moyamoya vessels being fed by the PCA group significantly increased in number with the severity of the steno-occlusive lesion in the ICA bifurcation, but they did not describe the network itself.

In most of the above publications the moyamoya anastomotic network is typically referred to as an “abnormal vascular network”, and presented as a haphazardly formed vascular tangle without defining its detailed anatomy. Moreover the angiographic descriptions are based on non-superselective injections. Therefore a detailed description of the angioarchitecture of the moyamoya anastomotic networks supplied by the Pcom and PCA branches is lacking.

Superselectivity in cerebral angiography has been implemented in our institution for 27 years and is routinely used as an additional diagnostic tool in selected cases. Its diagnostic yield and safety for children and adults has been proved for almost every aspect of cerebrovascular pathology¹⁵⁻²². Other groups have also reported on the role and safety of superselective catheterization for the endovascular approach to small calibre vessels with aneurysms, the evaluation of newly formed collaterals after revascularization procedures in children with moyamoya, even after catheterization of the vasospasm-prone middle meningeal and temporal arteries²³⁻²⁷.

Superselective microcatheterizations can offer more reliable information on micro-angioarchitecture and dynamic vascular collaterals, particularly important in moyamoya disease where many small-sized vessels are overprojected in less selective techniques. For surgical planning, the superselective injections provided not only additional information complementary to the clinical and haemodynamic evaluation but helped disclose the need for multiple revascularisation procedures, i.e. revascularisation not only for the MCA territory but also for the ACA and PCA territories.

Contrary to the general concept that moyamoya disease relatively spares the posterior circulation, stenotic lesions in the posterior circulation are observed quite often, in some series in a majority (75%) of patients²⁸. Our study confirmed the frequent involvement of the posterior circulation and the well-known collateral leptomeningeal network around the splenium

of the corpus callosum. This network can be fed either by the splenic artery or by more posteriorly running cortical branches of the distal PCA. In cases of steno-occlusive lesions of the PCA that vitiate the main retrosplenic leptomeningeal collateral network, other arterial branches including its dural branch, can offer a collateral supply to distal cortical ipsilateral or contralateral PCA territories by reconstituting the cortical vessels through durocortical connections and a fine leptomeningeal arterial network. This network connects dural tentorial branches with superficial arteries of the medial occipital cortex and is similar to the so-called ethmoidal moyamoya.

The posterior choroidal arteries at the level of the foramen of Monroe (Figures 4 and 5) were demonstrated to supply the pericallosal artery through midline septal transcallosal branches. The same vessels can also feed distal striatal arteries at the level of the angle of the lateral ventricles.

The connection of the moyamoya anastomotic network with the medullary system, described by Miyamoto et al.¹³ was confirmed by our study. Contrary to their assumptions on the limited role of the thalamoperforators in the supply of the medullary arteries, it was shown (Figure 2) that they could play a decisive role in the supply of the cortex in advanced stages of the disease of the anterior circulation. We also demonstrated that the thalamoperforating arteries of the P1 segment that irrigate the thalamus can feed the posterior portion of the basal ganglia through anastomoses with the distal segment of striatal arteries. These anastomotic connections must be located subependymally. Additionally, the same vessels can irrigate the rest of the thalamic territory through intrathalamic anastomoses with other thalamic arteries when steno-occlusive lesions of the parent vessel (PCA) deprived their normal anterograde supply.

The role that Pcom perforators and especially the tuberothalamic (premamillary) artery can play in the moyamoya anastomotic networks was demonstrated in two cases (Figures 7 and 8). While in other cases the collateral network was fed by the ventricular branches of the anterior choroidal, these cases demonstrated the importance of the tuberothalamic and other Pcom perforators which irrigated the distal segment of striatal arteries retrogradely, further contributing to the supply of the medullary system as well as the anterior cerebral artery through subependymal branches at the level of

the wall of the third ventricle. The case in Figure 6 shows that the posterior circulation offers collateral supply to the anterior circulation through not only the classic cortical connections, but also through basal anastomoses - in this case to the territory of the anterior choroidal - illustrating the significance of the collateral contribution of the posterior circulation in advanced stages of the disease in the anterior territory.

Our study has several limitations. First, only some of the examinations included superselective injections. Second, it is difficult to generalize in a disease where different patients exhibit different stages of the disease. Third, potential anatomic variations exist among different patients.

Conclusions

The literature is scant regarding the angioarchitecture of the posterior circulation in paediatric moyamoya disease. Even in publications describing the posterior circulation, only vague descriptions of an "abnormal vascular network" are found. From our study, moyamoya patients develop a predictable collateral network with complex anastomotic connections in the posterior circulation. High quality selective and superselective angiography of the posterior territory confirmed in part some earlier descriptions, but also revealed previously unreported connections within the moyamoya anastomotic network. This study helped clarify the anatomy of this anastomotic network including its feeding branches, course and recipient vessels.

Further studies of the collateral cerebral circulation, including a comparison with adult moyamoya, and cases of moyamoya syndrome, will further help define specificities of the collateral network in moyamoya patients. This work also has the potential to contribute to a new staging of the disease with clinical and therapeutic relevance and to shed more light on the response of normal cerebral vasculature under ischemic conditions.

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